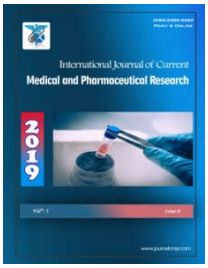
INTERNATIONAL JOURNAL OF CURRENT MEDICAL AND PHARMACEUTICAL RESEARCH
ISSN: 2395-6429, Impact Factor: 4.656
Available Online at www.journalcmpr.com
Volume 5; Issue 08(A); August 2019; Page No. 4457-4459
DOI: http://dx.doi.org/10.24327/23956429.ijcmpr2019081781



Research Article

# AN UNCOMMON ETIOLOGY OF ADRENAL GLAND MASS IN INFANT: PRIMARY HYDATID CYST

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**ARTICLE INFO**

***Article History:***
Received 6th May, 2019
Received in revised form 15th June, 2019
Accepted 12th June, 2019
Published online 28th August, 2019

***Key words:***
Cystic lesion; adrenal gland; hydatid disease; child

**ABSTRACT**

Hydatiddisease is caused by Echinococcus Granulosus, which is a larva endemic in many undeveloped areas. Hydatid disease is relatively uncommon in children. The liver and lungs are most commonly involved, while involvement of other sites in the body is unusual.
We report an uncommon case of hydatid disease located in the adrenal gland space.
This is a 13-year-old Tunisian boy, with left flank pain for three months previously without vomiting, transit disorders or jaundice and evolving in a context of conservation of the general condition and apyrexia. He was from a rural background and of low socioeconomic status. The contact with dogs or sheeps is confirmed. Clinical examination and preoperative imaging have suggested the diagnosis of neuroblastoma or ganglioneuroma or hydatid cyst of the adrenal gland. During the procedure, a hydatid cyst was found occupying the left adrenal gland.
Primary hydatidosis of the adrenal gland in children is extremely rare. Possible sources of infection include blood diffusion or local spread via lymphatic invasion. In the endemic areas, hydatid disease should be mentioned in the list of differential diagnoses of cystic lesions located around the adrenal gland space.



## INTRODUCTION

Hydatid disease is caused by Echinococcus Granulosus, which is a larva endemic in many undeveloped areas. It has maximum diffusion among people in poorer socioeconomic status so it poses a great problem to public health in countries where Echinococcus Granulosus is endemic. Tunisia is an endemic country.

Adrenal gland localization of the hydatid cyst is very rare, even in countries where hydatid disease is endemic, which may explain the difficulties of diagnosis. This location affects the child exceptionally.

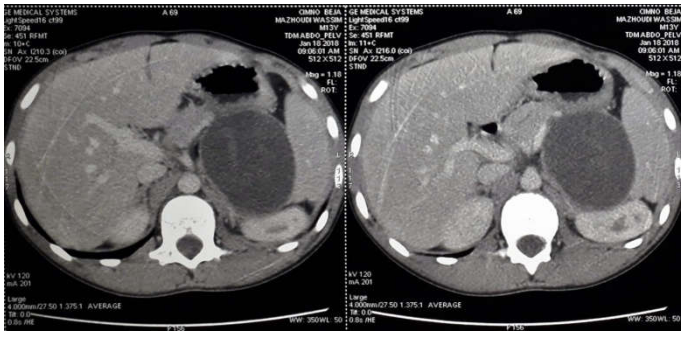
We report an observation of this localization occurring in a 13-year-old Tunisian boy and revealed by left flank pain. Through this observation and a review of the literature, we discuss the diagnostic difficulties and the modalities of the surgical treatment of this unusual localization of the hydatid disease.

## CASE REPORT

This is a 13-year-old boy from a rural area with no previous medical history. There was a history of animals 'contact, including dogs. He was hospitalized in our surgical paediatric department for recurrent left flank pain for three months previously without vomiting, transit disorders or jaundice and evolving in a context of conservation of the general condition and apyrexia.

The clinical examination was normal apart a palpable mass in the left renalangle.

CT scan abdomen showed a large well-defined oval shaped heavily mass at the left adrenal gland, measuring 6.7 × 6.1 × 7 cm (figures 1 and 2) the diagnosis of neuroblastoma or ganglioneuroma was evocated.



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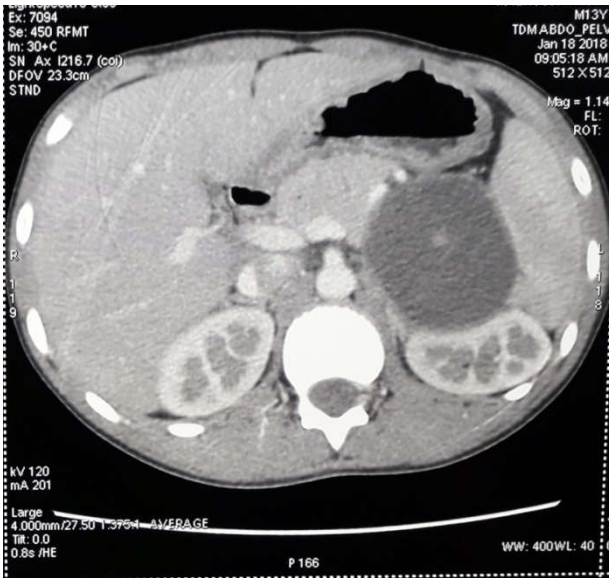
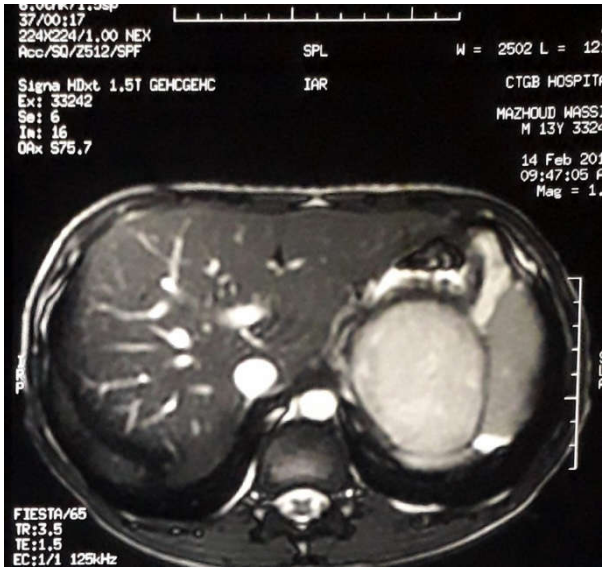
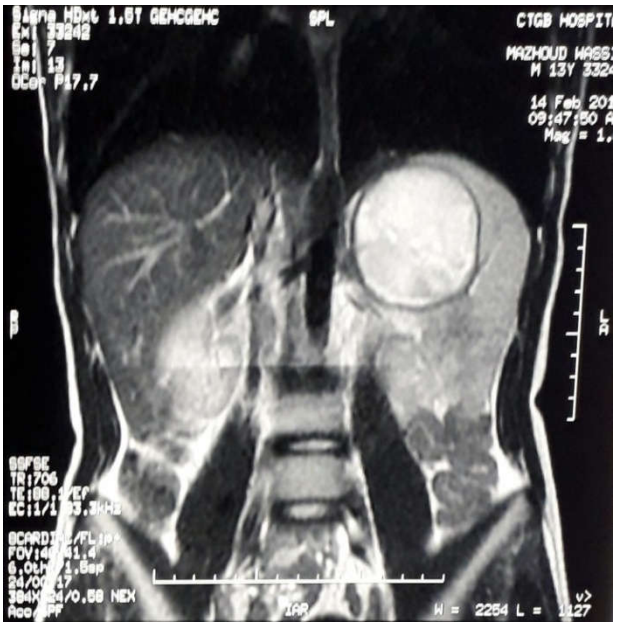



The abdominal MRI showed a heterogeneous cystic mass of the left adrenal gland. (Figure 3 and 4)

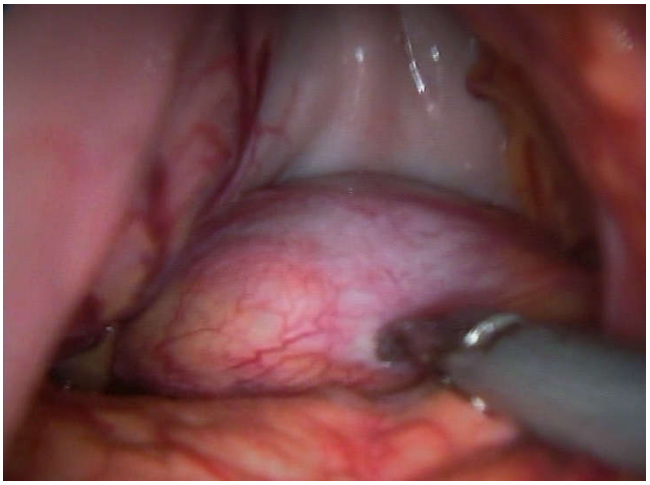



An ultrasound complement confirmed the existence of this cystic masse evoking hydatid cyst stage IV (classification by Gharbi).The remainder of the radiological assessment did not detect other hydatid localization, in particular hepatic or pulmonary.

Biology was normal: metanephrine, and normetanephrine were normal at 32ng/L (NR < 73), and 26 ng/L (NR <170) .
Hydatid serology (ELISA) was positive.

We decided to perform a surgical approach to this mass.We started a laparoscopic procedure that allows us to localize the mass. We could visualize the mass through the omentalbursa (figure5). We decided to convert to safely complete the operation .We used left subcostal incision.



Exploration found a cystic mass at the expense of the left adrenal gland lodge.

After protection of the surgical field and the abdominal wall by fields soaked in a scolicidal solution, draining of the mass was carried out, bringing a cloudyliquid, and a sterilization of the cyst with physiological saline serum. Then, we carried out an external drainage of the cystic cavity through a drain of Redon. Finally, the exploration of the peritoneal cavity did not objectify other localization. The liquid culture showed negative result.

Our patient was reviewed regularly at the consultation. With a follow-up of one year, ultrasound monitoring did not detect any hydatidrecurrence.

## DISCUSSION

Adrenal glands are one of the organs that are most rarely involved by hydatid cyst disease even in regions where the disease is endemic. As is the case for other hydatid cysts involving other organs, adrenal hydatidcyst is usually asymptomatic, usually being detected incidentally in radiological studies performed for other indications [2].

A multicentric study about diagnosis and treatment of abdominal hydatid cysts in children showed that the liver was the most common organ involved: in 69 cases it was the only organ involved and there were 2 kidney cyst cases, 8 spleens involved, 2 cases in broad ligament, 13 peritoneal cases and 26 cases of cyst located in great omentum. [5]

Adrenal hydatid cyst disease rarely becomes symptomatic, with most symptoms being related to the interaction of a cyst with adjacent organs and tissues (compression and inflammation). Particularly, inflammation causes signs and symptoms of peritoneal irritation. The most common symptoms are related to the gastrointestinal systems, such as flank pain, sense of fullness, constipation, and loss of appetite

[2]. It can be discovered in the context of an exploration of refractory arterial hypertension.

In our case, the hydatid cyst was detected after an abdominal pain and mass.

In paediatric population, cystic lesions of the adrenals may be grouped in three main types: “pure” cystic types (vascular or endothelial cyst), parasitic cysts and cystic part of an otherwise solid tumor (neuroblastoma, ganglioneuroma, pheocromocytoma, teratoma …) usually related to a process of necrosis or haemorrhage [4].

The radiological findings and a positive serology in our patient were highly suggestive of hydatiddisease.

Although serology is useful in the diagnosis of hydatid disease, a number of patients may have a negative test. In general, the sensitivity of the serological tests is determined by the location and state of the cysts. The indirect hem agglutination (IHA) test and ELISA have a sensitivity of 80% overall, but a negative serology does not exclude the diagnosis [3].

Hydatid cyst can be asymptomatic and need not any intervention except for doubt in the diagnosis and in large cyst causing mass effect. Treatment of adrenal hydatid, when indicated, is mainly surgical and by total cyst excision. Small asymptomatic non-functioning cysts are treated conservatively [3].

Total adrenalectomy may be considered when the cyst has completely destroyed the gland. Both laparoscopic resection of an adrenal hydatid and laparotomy are accepted surgical intervention.

Laparotomy nonetheless allows a better exploration of the peritoneal cavity.

Adjuvant Albendazole pre- and postoperatively reduced recurrences in hepatichydatidosis.

It may be argued that our patient infected hydatid cyst might have indicated dead parasites and hencedoes not require Albendazole therapy.

This patient’s history of contact with animals, the slowly progressive nature of his adrenal mass and positive serology together are highly suggestive of hydatid disease. The radiological findings are characteristic though not pathognomonic.

## CONCLUSION

Overall, isolated hydatid disease of the adrenals is rare.
The diagnosis should be suspected in all patients from or who lived in endemic areas. Surgical excision with either laparotomy or laparoscopic approach remains the intervention of choice in such cases. Adjunctive medical treatment improves the outcome and decreases the recurrence rate.

***Acknowledgment section:*** I would like to acknowledge with much appreciation the crucial role of the staff of Dr Kaabar, who gave the permission to use all required equipment and the necessary material to complete this work. A special thanks goes to Dr Hellal, who help me to assemble the parts and gave suggestion about the task. Last but not least, many thanks go to Dr Daib who has invested her full effort to supervise this work.

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**How to cite this article:**

Fatma Bchini *et al* (2019) 'An Uncommon Etiology of Adrenal Gland Mass in Infant: Primary Hydatid Cyst', *International Journal of Current Medical and Pharmaceutical Research,* 05(08), pp 4457-4459.

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